Endovascular Management of a Left Subclavian Artery Lesion Following Thoracoplasty for Bronchopleural Fistula and Empyema Secondary to *Aspergillus fumigatus*

Ricard Ramos,[a] Laura Rodríguez,[b] Josep Saumench,[c] Elena Iborra,[b] Marc Antoni Cairols,[b] and Jordi Dorca[d]

[a]Unitat d'Anatomia i Embriologia Humana, Servei de Cirurgia Toràcica, Hospital Universitari de Bellvitge, Universidad de Barcelona, L'Hospitalet de Llobregat, Barcelona, Spain
[b]Servei d'Angiologia i Cirurgia Vascular, Hospital Universitari de Bellvitge, L'Hospitalet de Llobregat, Barcelona, Spain
[c]Servei de Cirurgia Toràcica, Hospital Universitari de Bellvitge, L'Hospitalet de Llobregat, Barcelona, Spain
[d]Servei de Pneumologia, Hospital Universitari de Bellvitge, L'Hospitalet de Llobregat, Barcelona, Spain

Massive and/or recurrent hemoptysis is a clear indication for surgical treatment of pleuropulmonary aspergilloma, despite the incidence of postoperative morbidity and mortality. Thoracoplasty has been widely used for 20 years and is still indicated in these cases, following lobectomy, even though the procedure is not free of complications. We report the case of a patient who required thoracoplasty to treat a pleuropulmonary aspergilloma invading the chest wall. Subsequent placement of an aortic stent-graft was required due to tearing of the left subclavian artery.

Key words: *Pulmonary aspergilloma. Thoracoplasty. Thoracic aortic stent-graft.*

Manejo endovascular de lesión de arteria subclavia izquierda tras toracoplastia por fístula broncopleural y empiema secundario a *Aspergillus fumigatus*

La hemoptisis masiva y/o repetitiva es una indicación clara de tratamiento quirúrgico del aspergiloma pleuropulmonar, a pesar de la morbimortalidad posquirúrgica existente. La toracoplastia, muy utilizada hace 2 décadas, todavía tiene su indicación aquí, tras lobectomía, aunque no está exenta de complicaciones. Presentamos un caso de aspergiloma pleuropulmonar con invasión de pared torácica que requirió toracoplastia y posteriormente técnicas endovasculares aórticas por desgarro de la arteria subclavia izquierda.

Palabras clave: *Aspergiloma pulmonar. Toracoplastia. Endoprótesis de aorta torácica.*

Introduction

The type of pulmonary aspergillosis most frequently observed in pneumology and thoracic surgery departments is the saprophytic colonizing form known as pulmonary aspergilloma, which occurs in patients with cavitary sequelae and bronchiactasis.[1] Clinical findings range from absence of symptoms to massive hemoptysis. Blood-tinged sputum therefore indicates that aggressive treatment is required, though this therapeutic approach is currently under debate due to the postoperative morbidity and mortality found in all studies.[2,3] We report the case of a patient with recurring pulmonary aspergilloma who required lung resection, thoracoplasty, and endovascular exclusion, combined with carotid bypass surgery following tearing of the left subclavian artery.

Case Description

A 47-year-old man who smoked 2 packs per day and drank a moderate amount of wine presented with massive hemoptysis that required embolization of the left bronchial arteries. He was subsequently diagnosed with pulmonary mycobacteriosis due to *Mycobacterium kansasii*, with cavitary lesions of the left lung vertex. The patient followed a course of specific treatment with rifampicin, ethambutol, and isoniazid for 15 months, under management by the pneumology department; the clinical and radiological course was satisfactory. A few months after treatment had ended, the patient presented repeated episodes of minor hemoptysis associated with radiologic changes in the left upper lobe, which showed residual cavitation. Computed tomography of the chest revealed a spiculated cavitary mass in the left upper lobe, indicating a possible complex mycetoma. *Aspergillus fumigatus* was repeatedly isolated in sputum samples. Staining and cultures were negative for mycobacteria. A lung function study revealed a mild obstructive pattern and lung scintigraphy showed a slight reduction in perfusion of the left lung (38%, compared to 62% perfusion of the right lung). After the diagnosis of residual cavitation complicated by pulmonary aspergilloma, treatment was initiated with oral itraconazole at a dosage of 200 mg/12 h; the patient continued this regimen for the next 4 months. Intermittent blood-tinged sputum persisted despite treatment and *A fumigatus* continued to be isolated in sputum samples; no improvement was observed in the subsequent
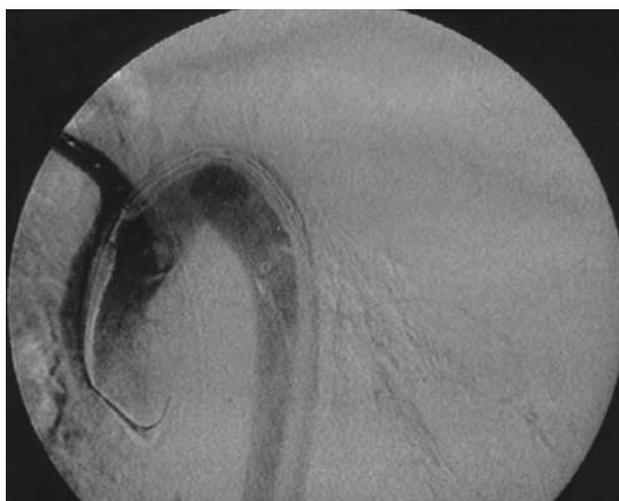
Figure 1. Insertion of the Medtronic Valiant thoracic stent graft through a port into the left common iliac artery.
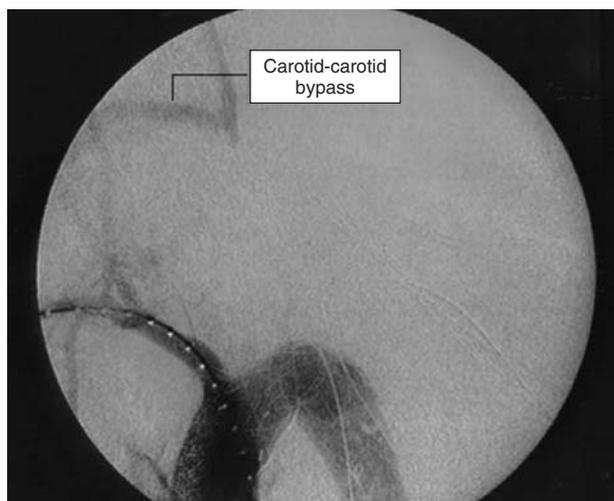

Figure 2. Angiography showing a permeable carotid-carotid bypass with no endoleaks.

radiologic follow-up. Surgery was finally indicated due to the persistent bleeding. An extrapleural left upper lobectomy was performed due to the presence of abundant apical pleural adhesions and invasion of both the pleura and the chest wall. The clinical course was marked by persistent air leaks and was subsequently complicated by a loculated posterior apical pleural empyema due to *A fumigatus*, leading to progressive clinical deterioration and scheduling of a new surgical intervention. Thoracoplasty was performed, with resection of the first 5 ribs. During surgery, the left subclavian artery was torn close to the aortic arch due to the large amount of fibrous tissue found throughout the posterior mediastinum. The artery was sutured but neither proximal nor distal dissection was not possible due to the abundance of chronic inflammation. Thrombosis occurred in the artery in the immediate postoperative phase. Acute ischemia of the left arm was treated with intravenous heparin sodium, with satisfactory clinical results, though the pulse from the left subclavian artery was not recovered. Arteriography showed complete obstruction of the subclavian artery and collateral circulation through the axillary and humeral arteries.

Fifteen days after surgery, the patient presented massive hemothorax with hemodynamic instability and an emergency repeat posterolateral thoracotomy was performed. Hemostasis was performed with Teflon stitches, due to the active hemorrhage in the aortic arch and the abundance of nondissectible fibrous tissue, and it was decided to perform elective endovascular surgery. Because the previously performed arteriogram showed a space of less than 1.5 cm between the left common carotid artery and the left subclavian artery, it was necessary to cover the ostium of the left common carotid artery prior to the bypass procedure in order to ensure the proximal anchor point for the stent.

Two consecutive surgical operations were performed. The first operation was a retroesophageal carotid-carotid bypass using an 8-mm ringed polytetrafluoroethylene graft, with ligature of the left proximal common carotid artery. Due to the small caliber of the left external iliac artery (70 mm), a left retroperitoneal approach was used to introduce a Valiant thoracic stent graft with Xcelerant delivery system (diameters, TF 4444 C 100) via a port through the left common iliac artery (Figure 1). The brachiocephalic artery was monitored by means of angiography using a right humeral catheter. The proximal anchor point covered the origin of the left common carotid artery. Angiography showed a permeable carotid-carotid bypass with no endoleaks (Figure 2).

Progress was satisfactorily after surgery. Bilateral carotid pulses were present, though electromyography confirmed paresis and anesthesia in the left arm due to injury of the brachial plexus.

The patient developed acute acathisia while on the ward, possibly due to the neuroleptic treatment, and required specific anxiolytic treatment. He died following progressive deterioration with reappearance of bilateral lung infiltrates and acute, progressive, and irreversible respiratory failure.

Discussion

Pulmonary aspergilloma is thought to be the most frequent clinical form of pulmonary aspergillosis, though there are no epidemiological studies to show that this is the case.[1] The fungus typically resides in already existing lung cavities—most often secondary to tuberculosis; it is less frequently found in bronchiectasis and giant bullae.[4] Although pulmonary aspergilloma is not an invasive process (it remains for years as a ball inside the lung cavity), it can, in some cases, erode the lung wall and give rise to the typical clinical manifestation of hemoptysis. Hemoptysis is usually mild and occurs in small quantities, but increases to the point of endangering the life of the patient. Approximately 10% of cases progress to massive hemoptysis.[5] This outcome is more common in patients who have previously suffered from tuberculosis. Our patient presented with massive hemoptysis due to erosion of the intercostal blood vessels, a situation described in the literature[6] and confirmed in our patient during a difficult lung resection, when the extrapleural fungal infection was observed and resection of the associated wall was initially rejected.

Surgery is currently the treatment of choice, in combination with antifungal drugs and/or embolization of tributary arteries when the patient presents hemoptysis, though opinion is divided, given the considerable postoperative morbidity and mortality. Segmental resection and particularly lobar resection are the most commonly used techniques; pneumonectomy is avoided whenever possible. Babatasi et al[3] highlight the risk of lung resections

due to abundant pleuropulmonary adhesions. Given these risks, some groups do not advocate prophylactic treatment of pulmonary aspergilloma.

As described in the literature[2] the most frequent complications are pleural. These include bronchopleural fistula and empyema, which often require aggressive treatment (eg, thoracoplasty). In our patient, the presence of extensive fibrosis and pleural infection led to a lesion of the left subclavian artery. The lesion was initially resolved, though the persistence of *A fumigatus* in the pleural fluid led to new bleeding that required a more definitive solution. After rejecting therapeutic solutions such as embolization of the subclavian artery[7] or placement of a subclavian stent[8] because of superinfection and friable tissue, it was decided that the most appropriate treatment was exclusion of the subclavian artery with an aortic stent.

The use of endovascular techniques to treat aortic stenoses and aneurysms has become widespread since the 1990s and results are satisfactory, especially in cases where conventional surgery involves high rates of morbidity and mortality.[8,9] Of great importance when using a stent is proper anchoring (requiring a length of >2 cm) to ensure the stability in the short and long term and to minimize the risk of endoleaks.[10] When the anchoring surface is too short, greater distance can be achieved by covering the left common carotid artery associated with the procedure by means of a carotid-carotid bypass—a technique commonly used to treat occlusions and aneurysms of the supraaortic arteries.[11]

The literature describes a risk of vertebrobasilar lesion following exclusion of the left subclavian artery (left dominance of the vertebral artery)[12] or subclavian steal syndrome[13-16]; this was unlikely in our patient's case due to the previous, well tolerated presence of occlusion of the left subclavian artery.

The risk of stent infection following aortic reconstruction must also be taken into account. This devastating complication of vascular surgery is associated with mortality rates of between 25% and 88%.[17] While the behavior of conventional materials in the context of infections has been widely studied, the behavior of stents in septic environments is still under investigation. In endovascular surgery, unlike conventional surgery, the mechanisms that protect the arterial tree against infection (circulating blood, including in the vasa vasorum) remain partially operative and this makes the stent more resistant to infection.[18,19] Some studies demonstrate good results when endovascular stents are used to treat mycotic pseudoaneurysms of the thoracic aorta.[12,20]

In conclusion, surgical treatment of pleuropulmonary aspergilloma must be performed with care, given the high rate of morbidity and mortality. As shown in this case, it may be necessary to undertake surgical treatment that is neither usual in this disease nor standard in thoracic surgery departments. Endovascular techniques are an effective therapeutic option in cases of acute hemorrhage.

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